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Case Report

Syringocystadenoma Papilliferum of Eye Lid: A Case Report and Review of Literature in a Tertiary Eye Hospital, Nigeria

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Abstract

A 58-year-old male with a one-year history of lower medial eyelid swelling and no other ocular and systemic abnormalities was examined. The examination revealed a medial bluish firm left lower eyelid mass. Subsequently, he had an *in toto* excisional biopsy of a cystic mass, which was confirmed histopathologically to be syringocystadenoma papilliferum. A higher level of suspicion by the ophthalmologist and the histopathologist plays a vital role in the management of this tumour.

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1. Introduction

Syringocystadenoma papilliferum (SP) of the eyelid is a rare benign tumour of the Moll's glands equivalent to the sweat gland first reported in 1917 by John Stokes [1]. The tumour is not so common in the eyelid, and is often misdiagnosed as Cyst, BCC, or SCC [2]. Hence, histopathology evaluation is needed to confirm the diagnosis.

Syringocystadenoma papilliferum is mostly a childhood tumour [3] with 50% reported at birth and 15–35% presented at puberty [4]. However, it has been known to be associated with other benign tumours such as viral warts, nevus sebaceous, linear nevus verrucous, nevus comedonicus, apocrine poroma, apocrine hidrocystoma, tubulopapillary hidradenoma, hidradenoma papilliferum and papillary eccrine adenoma [5]. Syringocystadenoma papilliferum can rarely transform into BCC, as has been reported [4].

A thorough review of the literature in Google Scholar, *African Journals Online (AJOL)* and Medline/PubMed yielded 17 cases of SP of the eyelid. We hereby report another interesting case of SP of the eyelid.

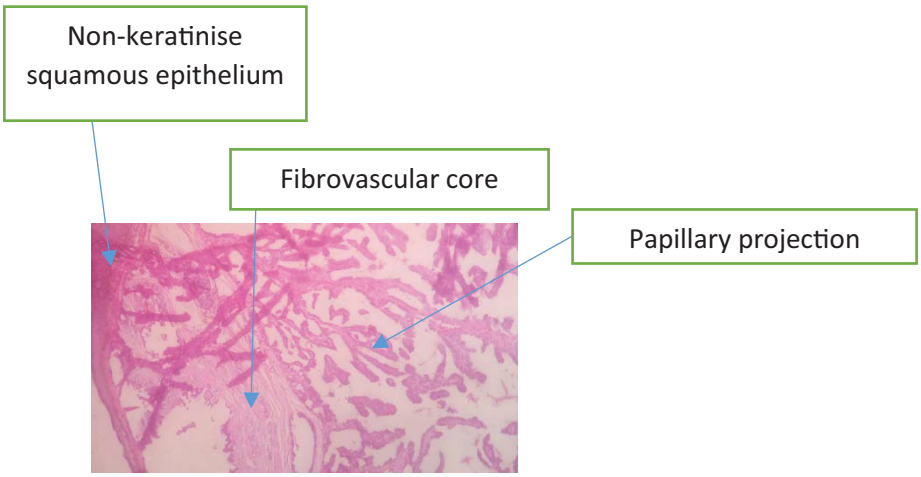

Figure 1: Microscopy section shows projected papillary, plasmacytic infiltrates and non-keratinised epithelium.

2. A Case Report

A 58-year-old male with a non-progressing right lower eyelid swelling of one-year duration, unassociated with pain, bleeding, itching and eye lashes loss was examined. There is no preceding history of trauma and weight loss. Visual acuity (VA) in both eyes was 6/6.

Physical examination revealed medial bluish firm Right Eye (RE) swelling measured 1.2cm x 0.6cm. Other ocular findings were normal in Both Eye (BE). There is no significant peripheral lymphadenopathy. The mass was excised *in toto*, and the wound was closed primarily.

3. Histopathological Report

Gross examination showed a round to oval mass 10mm x 5mmm.

Microscopic examination of tissue sections shows an acanthotic non-keratinised squamous epithelium overlying a cystic lesion composed of intra-cystic papillary structures lined by double layer of inner columnar and outer cuboidal cells (Figure 1) with decapitation secretion seen within the luminal spaces. The fibrovascular cores show plasmacytic infiltrations with decapitation secretion within the luminal spaces.

4. Discussion

Syringocystadenoma papilliferum is a rare hamartoma benign tumour of the eyelid first reported [1] in 1917, believed to arise from sweat gland and mostly seen in head and neck region [1]. However, the occurrence of the tumour in other part of the body have been reported. From looking into the literature through Google Scholar, Medline/PubMed and *African Journals Online (AJOL)*, 17 cases of eyelid SP have been reported as of today. The highest number was by Barbarino et al. in 2009, who reported 14 number of cases, with age ranging 8–82years [6]. Our case was a 58-year old similar to the age reported by Behera & Chatterjee [2]; however, older than the age reported by Rao et al. [3] and Jakobiec et al. [7] . A clinical differential diagnosis of this lid mass in our patient includes nevus sebaceous, hidradenoma, dermoid cyst, BCC, SCC and keratoacanthoma. However, BCC and SCC are unlikely because of the age of our patient. Keratoacanthoma is a rapid-growth tumour, however, the non-rapid growth nature and age in our patient was not in favour of Keratoacanthoma. The histopathological diagnosis of SP by Jakobiec et al. [7] were all seen in our patient (cystic spaces within dermal, the dermis lined by non-keratinised epithelium, papillary projection and prominent plasmacystic infiltration). There are still arguments pertaining to the origin of SP: while some authors believe its apocrine origin, others call it eccrine gland origin [8]. In our patient, apart from the eyelid swelling no other ocular symptoms were noticed; however, there were previous reports of association of pruritic [9], verrucous and hyperkeratotic surface lesion [6] with SP. Syringocystadenoma papilliferum is known to be associated with many other benign tumours such as apocrine cystadenoma [6], condyloma acuminatum [10], trichilemmoma [11], hidradenoma [12], trichoblastoma [13], verrucous cyst [14] and tubular apocrine adenoma [15], while our patient was associated with apocrine hydrocytoma.

The malignant transition into BCC [2], ductal carcinoma and metastatic adenocarcinoma [16] have been reported in SP of other part of the body but none in the eyelid. The main treatment of SP of eyelid is excisional biopsy that was done in our case; however, carbon dioxide laser [17] and Mohs micrographic surgery [18] have been used successfully in the treatment of SP. Two cases of recurrences of eyelid SP following surgical excisional have been reported in the literature [19].

5. Conclusion

Syringocystadenoma papilliferum of eyelid still remains a rare benign tumour that can be easily misdiagnosed as cyst, BCC and SCC; hence higher index of suspicion by all practising ophthalmologist and histopathologist plays an important role in its diagnosis.

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